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Linking Anxiety and Insistence on Sameness in Autistic Children: The Role of Sensory Hypersensitivity

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Abstract Sensory hypersensitivity and insistence on sameness (I/S) are common, co-occurring features of autism, yet the relationship between them is poorly understood. This study assessed the impact of sensory hypersensitivity on the clinical symptoms of specific phobia, separation anxiety, social anxiety and I/S for autistic and typically developing (TD) children. Parents of 79 children completed questionnaires on their child's difficulties related to sensory processing, I/S, and anxiety. Results demonstrated that sensory hypersensitivity mediated 67% of the relationship between symptoms of specific phobia and I/S and 57% of the relationship between separation anxiety and I/S. No relationship was observed between sensory hypersensitivity and social anxiety. These mediation effects of sensory hypersensitivity were found only in autistic children, not in TD children.

Keywords Autism spectrum disorder · Hypersensitivity · Anxiety · Insistence on sameness · Sensory processing

Introduction

Nearly 75 years after Kanner (1943) first described sensory difficulties in autism spectrum disorder (ASD), the role that atypical sensory processing plays within the etiology of the disorder remains poorly understood.

Karen R. Black and Ryan A. Stevenson contributed equally to this publication.

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Upwards of 88% of autistic children[1] experience abnormalities due to sensory sensitivities (Baum et al. 2015; Leekam et al. 2007; Tomchek and Dunn 2007), which are thought to present in early toddlerhood and remain stable throughout the elementary school years (Green et al. 2012, 2015; McCormick et al. 2015). Diagnostic criteria in the DSM-5 criteria include abnormal sensory behavior as only a component within the over-arching construct of restricted interests and repetitive behaviors (RRBs) (APA 2013), yet researchers report links between hypo- and hyper-sensitivity and RRBs (Boyd et al. 2010; Gal et al. 2010; Lidstone et al. 2014), as well as between sensory sensitivity and anxiety (Green et al. 2012; Lane et al. 2012; Lidstone et al. 2014; Mazurek et al. 2012; Tseng et al. 2011). Although anxiety is not considered a core feature of autism, co-morbidity is reported in at least 40% of autistic children (Simonoff et al. 2008; Van; Steensel et al. 2011; White et al. 2009), with findings indicating long term associations with depression, aggression, hyperactivity and self-injury (Kim et al. 2000; Mazzone et al. 2013; Segers and Rawana 2014; von Klitzing et al. 2014).

Over responsiveness to sensory stimulation (*sensory hypersensitivity*) may play a key role in the development of anxiety in autistic children (Green and Ben-Sasson 2010; Green et al. 2012; Lane et al. 2012; Lidstone et al. 2014). A directional pattern has been observed whereby autistic adolescents demonstrate increased sensory hypersensitivity after simultaneous presentation of uncomfortable audio or visual stimuli such as hearing a vacuum cleaner (Green et al. 2015). By the same token, intense phobias are hypothesized to originate following an overwhelming sensory experience (Green and Ben-Sasson 2010). Furthermore, sensory hypersensitivity in autistic toddlers has been found to predict later development of anxiety, while anxiety did not predict changes in sensory hypersensitivity (Green et al. 2012).

Correlations between sensory hypersensitivity and insistence on sameness behaviors such as behavioral inflexibility and narrow interests (I/S), have also been observed (Lane et al. 2014; Leekam et al. 2011; Lidstone et al. 2014; Kerns et al. 2014; Tseng et al. 2011). Linked with increased levels of anxiety, the rigidity associated with I/S has been theorized to exacerbate and compound distress (Green and Ben-Sasson 2010). However, it has also been suggested that I/S may function as a coping mechanism (Lidstone et al. 2014; Rodgers et al. 2012; Wigham et al. 2015). Consequently, sensory hypersensitivity may help to explain the co-occurrence of both anxiety and I/S in autistic children (Green and Ben-Sasson 2010; Lidstone et al. 2014).

To date, only two studies have been conducted investigating the nature of the relationships between sensory hypersensitivity, anxiety, and I/S, with each proposing different models (Lidstone et al. 2014; Wigham et al. 2015). In the first study, sensory hypersensitivity (measured as sensory avoiding and sensory sensitivity) mediated the relationship between anxiety and I/S (Lidstone et al. 2014); while in the second, anxiety (with intolerance of uncertainty) mediated the relationship between sensory hypersensitivity and I/S (Wigham et al. 2015). Thus, while the directionality of the relationships is unclear based on the available evidence, there is converging evidence for a three-way relationship between sensory hypersensitivity, anxiety, and I/S in autistic children.

Importantly, both studies used a measure of total anxiety, leaving the impact of anxiety subtypes unexplored. The total anxiety construct is commonly used in survey measurement; however, numerous studies indicate high rates of difficulties experienced by autistic children due to particular anxiety subscales (Kerns et al. 2014; Kim et al. 2000; Muris et al. 1998; Rodgers et al. 2016; Simonoff et al. 2008) even while only a minimal association is often observed between I/S and *total* anxiety (Gotham et al. 2013). Thus, a single, overall measure of anxiety may lack the sensitivity to capture nuanced relationships between anxiety, hypersensitivity and I/S in autism. Based on previous experimental and theoretical research, we hypothesize that three anxiety subtypes in particular are likely to be related to sensory hypersensitivity and/or I/S – (1) specific phobia, (2) separation anxiety, and (3) social anxiety.

Specific phobia has been shown to be positively related to hypersensitivity (Kerns et al. 2014), and the *fear of physical injury* subscale, a related construct from the Spence Children's Anxiety Scale (SCAS), has been shown to significantly correlate with I/S (Rodgers et al. 2012). Theoretically, it has been hypothesized that specific phobia may be driven by sensory hypersensitivity in ASD given that specific phobias in ASD are frequently associated with loud noises or loud environments (e.g. crowded places) (Gjevik et al. 2011; Leyfer et al. 2006; Mukaddes and Fateh 2010; Russell et al. 2005).

Similarly, *separation anxiety* has been linked to I/S in multiple studies, including in both non-ASD (Boelen et al. 2014) and ASD samples (Rodgers et al. 2012). In

[1] While researchers and clinicians often feel more comfortable using person-first language such as "individuals with autism," autistic individuals have endorsed identity-first language that incorporates autism as a component of their identity over person-first language (61% vs. 28%; Kenny et al. 2015). While autistic individuals themselves bear the full ability to choose which terminology is used, these preferences also coincide with parents of autistic children (51% vs 22%; Kenny et al. 2015) and self-advocates (Sinclair 1999, Brown & ASAN, 2016; IFA 2016). As such, we will respect this preference and use this language throughout this manuscript.

Table 1 Participant exclusions

Reason	ASD N	TD N
Missing complete measure	2	2
Failure to provide document showing official diagnosis	2	–
Failure to exceed criteria for sample	3	5
Self-report of psychiatric disorder[a]	–	4
Self-report of neurological disorder[b]	1	–
Outliers—specific phobia	1	–
Outliers—separation anxiety	–	1
Outliers—social anxiety	1	–
Outliers—hypersensitivity	–	2
Outliers—FSIQ2	1	–
Total	11	14

[a]ASD participants were not excluded for report of co-morbid psychiatric disorders

[b]I.e. Epilepsy, brain damage, or other documented neurological issue

the latter, measurements were taken on a wide range of anxiety subscales using the SCAS, and correlated with the I/S factor score from the Repetitive Behaviours Questionnaire. Not only was separation anxiety significantly correlated with I/S in ASD, but it was also the anxiety subscale that showed the strongest correlation (Rodgers et al. 2012). The relationship between separation anxiety and sensory hypersensitivity in ASD has, to our knowledge, not been empirically tested, but will be explored in the current study.

Finally, *social anxiety* has yet to be measured with the SCAS in the context of sensory sensitivity and I/S, however, socio-emotional processing difficulties in adolescents with ASD have been shown to lead to heightened activation in the amygdala, pre-frontal cortex and striatum (Weng et al. 2011). Similarly, communication impairments have been theorized to result in a constant state of confusion and threat awareness for adolescents with ASD (Tupak et al. 2014). For example, recent fMRI findings demonstrated that amygdala over-responsivity in children with ASD was directly related to multiple co-occurring sensory experiences (Green et al. 2015).

Previous questions raised in the literature as to the greater implications of specific subtypes of anxiety in ASD suggest that associations between social anxiety, specific phobia and separation anxiety may contributed differently to hypersensitivity and I/S. Furthermore, many studies in the literature noted the lack of available research including a non-clinical control group. Therefore, in the current study, we aimed to address these gaps in our understanding of the relationships between sensory hypersensitivity, anxiety subtypes, and I/S in autistic children. We explored:

1. Whether sensory hypersensitivity influences the relationship between subtypes of anxiety and I/S in autism, particularly: *Specific phobia, social anxiety, and separation anxiety*.
2. Whether these relationships differ between autistic (ASD) and typically developing (TD) children.

For autistic children, we hypothesized that specific phobia, social anxiety, and separation anxiety would each individually predict I/S behaviors, and that these relationships would be mediated by sensory hypersensitivity. We further predicted that such relationships would not be observed, or observed to a significantly lesser extent, in age-matched, TD children.

Materials and Methods

Participants and Procedure

Parents of 104 children (ASD = 50, TD = 54) completed self-report measures while their children participated in diagnostic interviews. The study took place at the University of Toronto and York University and was approved by research ethics boards at both locations. Participants were recruited through local community organizations in Toronto, ON. A subset of participants (n = 25) were excluded prior to data analysis for issues including missing data (4), TD children reporting psychiatric diagnoses or exceeding ASD cutoffs (9), and autistic children failing to meet diagnostic criteria (5), reporting neurological disorders (1), or exclusion of outliers following analyses (6). For a complete breakdown of exclusions by reason and groups, see Table 1. Our final sample thus included 79 children: 39 autistic children and 40 TD children. Detailed demographic information of included participants are outlined in Table 2.

Diagnostic Measures

Clinical diagnosis of autism was confirmed by receipt of a child's original diagnostic report, administration by a research-reliable experimenter of the Autism Diagnostic Observation Schedule, versions 1 or 2 (Lord et al. 2012), and completion of the Autism Quotient: Child (AQ-C; Auyeung et al. 2008). Data from the AQ-C was also examined as a measure of symptom severity; higher scores have been shown to represent a higher expression of overall behaviors characteristic of autistic children (Auyeung et al. 2008; Paquette-Smith et al. 2014). Verbal and performance IQ were estimated for all children with

Table 2 Demographic and symptomatic variables across autistic (ASD) and typically developing (TD) children

	ASD (n = 39)			TD (n = 40)			95% CI	$t_{(77)}$	p	d
	Mean	SD	Range	Mean	SD	Range				
Demographic										
Gender								$\chi^2_{(1)}$: 11.1	0.001[a]	−0.37[b]
Age	12.1	2.6	7–17	11.0	3.0	7–18	[−0.2 2.3]	1.6	0.104	0.39
VIQ	45.0	13.3	20–67	57.9	8.4	39–78	[−17.9–7.9]	−5.1	<0.0001	1.16
PIQ	47.3	12.4	20–63	51.9	7.1	38–66	[−9.1–0.1]	−2.0	0.047	0.45
Symptomatic										
AQ	97.9	13.6	69–127	49.6	11.7	34–68	[42.7 54.0]	16.9	<0.0001	3.8
Hypersensitivity	28.8	10.5	12–55	12.6	5.0	0–21	[12.5 19.9]	8.8	<0.0001	1.96
I/S	11.3	3.7	6–18	7.0	1.1	6–10	[3.1 5.5]	7.0	<0.0001	1.57
Specific phobia	5.1	3.7	0–14	3.8	2.7	0–10	[−0.1 2.7]	1.8	0.075	0.40
Separation anxiety	3.9	3.4	0–12	2.9	2.4	0–9	[−0.2 2.4]	1.6	0.111	0.34
Social anxiety	4.4	3.5	0–12	3.9	2.7	0–9	[−0.8 2.0]	0.8	0.426	0.16

VIQ Verbal IQ (WASI-II), *PIQ* Performance IQ (WASI-II), *AQ* Symptom severity (Autism Quotient), *I/S* Insistence on sameness

[a & b]phi coefficient significance & effect size

the vocabulary and matrices subtests of the Wechsler Abbreviated Scale of Intelligence-II (WASI-II; Wechsler 2011).

Repetitive Behavior Questionnaire 2 (RBQ-2; Leekam et al. 2007)

The RBQ-2 is a 20-item caregiver-report questionnaire that examines the frequency and severity of RRBs. Respondents rate child behavior within the last month on a 3-point scale (*1—never/rarely* to *3—marked/notable*). Lidstone et al. (2014) derived a two-factor grouping for I/S and repetitive motor movements (RMM), excluding sensory items to control for over-inflation across measures. As the scale previously demonstrated good internal consistency for both I/S and RMM (∝ = 0.76/.83) for autistic children, we used the I/S factor, summing I/S items to create the I/S score.

The Short Sensory Profile (SSP; McIntosh et al. 1999)

The SSP is a brief, optimized version of Ermer & Dunn's full-length Sensory Profile (SP) questionnaire (1998), identifying how sensory processing difficulties relate to a child's participation in daily activities. The original SP provides two factors for assessing sensory sensitivities (*avoiding* and *sensitivity*), rated on a 5-point Likert scale (*never to always*) however, many of the items in the avoiding cluster are more closely related to anxiety and emotional regulation than sensory abnormalities (i.e.: *expresses feeling like a failure; has definite, predictable fears; has temper tantrums*). The SSP addressed this issue by omitting the emotion-related items, however, clusters of items could still have been combined due to close thematic overlap. For example, between *auditory filtering* and *visual/auditory sensitivities* or between *tactile sensitivity* and *taste/smell sensitivity* (e.g.: *"limits self to particular food textures"*). Although both the SP and the SSP have displayed strong validity discriminating between children with and without sensory abnormalities (>95%), neither tool provides a specific measure of *hypersensitivity*. Based on recent recommendations to tailor existing instruments to improve measurement of symptom expression in ASD (Magiati et al. 2016; White et al. 2015), we extracted a hypersensitivity construct from a factor analysis of the SSP by Tomchek, Huebner, & Dunn (2014) by selecting question items from the SSP demonstrating the highest factor loading, and best conceptual fit for the model. Eleven items with the highest factor loadings were included, combining descriptors from both original SP quadrants of *sensory sensitivity* and *sensory avoiding* (Ermer and Dunn 1998). Our hypersensitivity factor showed good internal consistency for both the ASD (∝ = 0.85) and TD groups (∝ = 0.74). Items are listed in Table 3. Items were scored in line with the original SP such that greater levels of hypersensitivity are indicated by higher scores.

Spence Children's Anxiety Scale: Parent Report (SCAS-P; Spence 1998)

The SCAS was developed to assess the symptoms of six different types of child anxiety within the general population: *panic/agoraphobia, separation anxiety, social anxiety, fears of physical injury, obsessive–compulsive disorder, generalized anxiety*. Symptoms are rated on a 4-point Likert scale (*never to always*) and scores are derived by

Table 3 Hypersensitivity factor

Item	SP quadrants	SSP factor loading
1. Rejects tastes/food smells typical of children's diets	SS	0.853
2. Limits self to certain food textures	SS	0.852
3. Unproductive with background noise	AV	0.718
4. Reacts aggressively to being touched	SS	0.637
5. Holds hands over ears to protect from sound	AV	0.635
6. Reacts strongly to loud noises	AV	0.620
7. More bothered by bright lights than others	SS	0.596
8. Rubs or scratches a part of the body when touched	SS	0.586
9. Anxious when standing close to others	SS	0.551
10. Distressed during grooming	SS	0.460
11. Looks away from tasks to notice all actions in room	SS	0.457

SP Sensory profile, *SSP* Short sensory profile, *SS* sensory sensitivity, *AV* sensory avoiding

Table 4 Specific phobia subscale

Item	Factor items
1. My child is scared of the dark	O
2. My child is scared of dogs	O
3. My child is scared of going to the doctor or dentist	O
4. My child is scared of heights	O
5. My child is scared if (s)he has to travel in the car, bus or train	P
6. My child is scared of insects or spiders	O
7. My child is afraid of being in small, closed spaces, like tunnels or small rooms	P

O Original fears of physical injury subscale items, P Panic subscale items common to specific phobia in the ADIS-IV

summing item totals within each subscale, and an overall total of all items. The individual subscales have demonstrated good internal consistency (∝ = 0.47–87) and test–retest reliability (∝ = 0.60–82) with TD children (Spence 1998; Arendt et al. 2014). Based on our *a priori* hypotheses, our goal was to examine three anxiety subtypes in autistic children: *Social anxiety, specific phobia, and separation anxiety*. As the SCAS does not include a subscale for specific phobia, and recent analyses of the factor structure indicate concerns regarding the validity of all six subscales (Glod et al. 2017; Jitlina et al. 2017), *fears of physical injury* was adapted to allow for phobia symptom measurement. We followed the outline for specific phobia in the *Anxiety Disorders Interview Schedule for DSM-IV* (Silverman and Albano 1996) and added two additional items from the SCAS found in the panic subscale. The new 7-item specific phobia subscale (Table 4) demonstrated an increase in consistency within both samples (ASD/TD ∝ = 0.60) relative to previous TD group norms for the fears of physical injury subscale (previous ∝ = 0.50; (Arendt et al. 2014; Glod et al. 2017), and using a polychoric correlation specific for ordinal data (Gadermann et al. 2012), showed Ordinal Cronbach's α-values of 0.76, 0.75, and 0.68 for the total sample, ASD, and TD, respectively.

Analysis

We examined group comparisons for all dependent variables with a set of t-tests, followed by pairwise correlations. Measures of anxiety subtypes that differed between the ASD and TD groups in their relationship to either sensory hypersensitivity or I/S were further examined in a subsequent mediation analysis with hypersensitivity and I/S, using a bootstrap procedure with 5000 resamples (Preacher and Hayes 2004). In all regressions, predictor variables were centered around their mean. Missing items for 14 participants (ASD = 3, TD = 11) were assessed to confirm that the pattern of missing data was random, and then were replaced using multiple imputation. An average of 2% of the total items were imputed per individual, with none required for the hypersensitivity factor.

Results

Demographics (Table 1)

T-tests were conducted to assess differences across ASD and TD groups for demographic variables (age, verbal IQ t-scores, performance IQ t-scores) and a χ^2 test to assess gender differences. ASD ($M=12.08$ years, $SD=2.63$) and TD ($M=11.03$ years, $SD=3.03$) groups did not significantly vary on age ($t_{(77)}=1.64$, $p=0.10$, CI [−0.22, 2.32], $d=0.37$), but in line with ASD prevalence, the ASD group was more heavily weighted towards males (ASD = 76.9%, male, TD = 40% male). Furthermore, the groups significantly differed for verbal IQ ($t_{(77)}=-5.15$, $p<0.001$, CI [−17.86, −7.91], $d=1.17$) and performance IQ ratings ($t_{(77)}=-2.0$, $p=0.05$, CI [−9.1 −0.1], $d=0.46$), thus we controlled for these differences with two regressions including gender, verbal IQ and performance IQ predicting (1) I/S ($F_{(38)}=0.95$, $p=0.43$) and (2) hypersensitivity ($F_{(38)}=1.00$, $p=0.40$). Neither model was significant.

Between-Group Relationships: ASD Symptomatology

A second set of t-tests compared differences in symptomatology across ASD and TD groups. All correlations were corrected for multiple comparisons using a Holm-Bonferroni correction. Results revealed significant differences between groups for symptom severity (ASD: $M=97.95$, $SD=13.63$, TD: $M=49.60$, $SD=11.69$; $t_{(77)}=16.94$, $p<0.0001$, CI [42.67, 54.03], $d=3.86$), hypersensitivity (ASD: $M=28.77$, $SD=10.49$, TD: $M=12.58$, $SD=4.97$; $t_{(77)}=8.80$, $p<0.0001$, CI [12.53, 19.86], $d=2.01$), and I/S (ASD: $M=11.28$, $SD=3.69$ TD: $M=6.98$, $SD=1.12$; $t_{(77)}=7.05$, $p<0.0001$, CI [3.09, 5.52], $d=1.60$). Examination of anxiety subscales showed that only *specific phobia* trended towards significantly higher levels in the ASD group ($t_{(77)}=1.80$, $p=0.07$, CI [−0.14, 2.74], $d=0.41$). Despite numerical mean differences, no significant differences were found between groups for *social anxiety* (ASD: $M=4.44$, $SD=3.46$, TD: $M=3.88$, $SD=2.73$; $t_{(77)}=0.80$, $p=0.43$, CI [-0.83, 1.96], $d=0.18$) or *separation anxiety* (ASD: $M=3.95$, $SD=3.44$, TD: $M=2.88$, $SD=2.40$; $t_{(77)}=1.62$, $p=0.11$, CI [−0.25, 2.40], $d=0.37$).

Correlational Relationships (Fig. 1)

Hypersensitivity and I/S were examined individually for pairwise correlations with each other and with each of the three anxiety subtypes: *specific phobia, social anxiety, separation anxiety*. All correlations were corrected for multiple comparisons using a Holm-Bonferroni correction. Significant correlations were only observed in the ASD group. Hypersensitivity demonstrated a strong correlation with I/S ($r=0.71$, $p<0.0001$), as well as with specific phobia ($r=0.54$, $p<0.0001$) and separation anxiety ($r=0.53$, $p<0.0001$). The relationship between hypersensitivity and social anxiety did not reach significance ($r=0.26$, $p=0.12$). In the TD groups, hypersensitivity was not significantly correlated with I/S ($r=0.19$, $p=0.24$), specific phobia ($r=0.11$, $p=0.50$), separation anxiety ($r=-0.25$, $p=0.12$), or social anxiety ($r=0.07$, $p=0.67$). Direct comparisons of sensory hypersensitivity, I/S, and individual anxiety subscales were also conducted. The ASD group showed stronger correlations than their TD peers between hypersensitivity I/S ($z=2.97$, $p=0.003$), as well as between hypersensitivity and both specific phobia ($z=2.10$, $p=0.03$) and separation anxiety ($z=3.57$, $p=0.0004$), but not social anxiety ($z=0.84$, $p=0.40$).

I/S, however, was positively correlated in ASD with all three subtypes of anxiety: specific phobia ($r=0.50$, $p=0.001$), separation anxiety ($r=0.54$, $p<0.0001$), and social anxiety ($r=0.33$, $p=0.04$). Again, no significant relationships were found in the TD group with specific phobia ($r=0.25$, $p=0.12$), separation anxiety ($r=0.11$, $p=0.50$), or social anxiety ($r=0.23$, $p=0.15$). Direct comparisons of I/S and individual anxiety subscales were also conducted. The ASD group showed a stronger correlation than their TD peers between I/S and separation anxiety ($z=2.11$, $p=0.03$) but not specific phobia ($z=1.28$, $p=0.20$) or social anxiety ($z=0.46$, $p=0.65$).

As specific phobia and separation anxiety showed correlations with both hypersensitivity and I/S in the ASD group, we further examined their relationships in ASD using mediation analyses. The lack of similar significant correlations (see Fig. 1) mathematically precluded the possibility that mediation effects occurred in the TD group (for analyses, see supplemental materials).

Mediation: Specific phobia (Fig. 2)

Overall, specific phobia was related to I/S ($c=0.50$, $SE=0.17$, 95% CI [0.15, 0.80], $p<0.0001$). The mediation analysis further revealed a significant indirect pathway from specific phobia to I/S through hypersensitivity ($ab=0.33$, $SE=0.11$, 95% CI [0.10, 0.55], $p<0.001$), with hypersensitivity mediating 67% of the total effect. However, the direct path accounting for the contribution of specific phobia to I/S, independent of hypersensitivity, was not significant ($c'=0.16$, $SE=0.14$, 95% CI [−0.03, 0.51], $p=0.12$).

Two additional models were examined for the three variables, testing alternative mediators in the place of hypersensitivity. The first of these included specific phobia as a mediator between sensory hypersensitivity and I/S, and was not significant ($ab=0.03$, $SE=0.03$, 95% CI [−0.01, 0.11], $p=0.11$). The second included I/S as a mediator

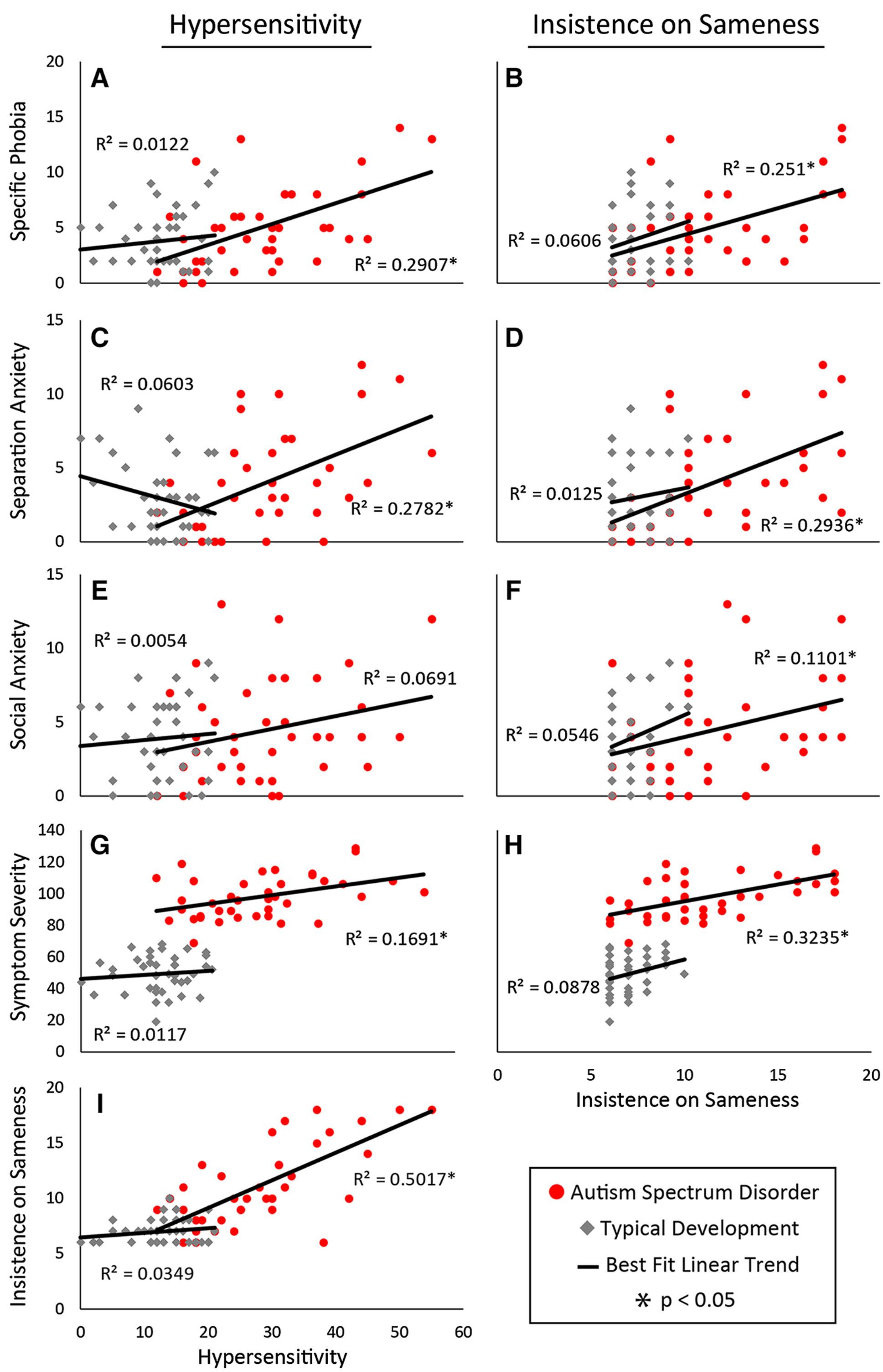


Fig. 1 Pairwise correlations between anxiety subtypes, hypersensitivity and I/S for autistic and typically developing groups. *Indicates significance after a Holm-Bonferroni correction for multiple comparisons

between sensory hypersensitivity and specific phobia, and was also not significant (ab = 0.06, SE = 0.04, 95% [−0.02, 0.13], p = 0.12). These additionally tested models provide evidence for the specific directionality of the relationships described in our original model (Fig. 2).

Mediation: Separation Anxiety (Fig. 3)

The mediation analysis of the relationship between separation anxiety, hypersensitivity and I/S revealed similar results to those observed with specific phobia in the model.

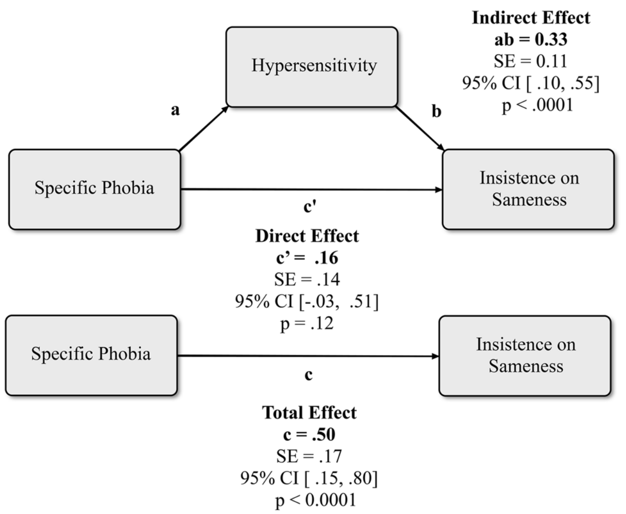


Fig. 2 Mediation model testing the effect of hypersensitivity on the relationship between specific phobia and I/S (ASD group)

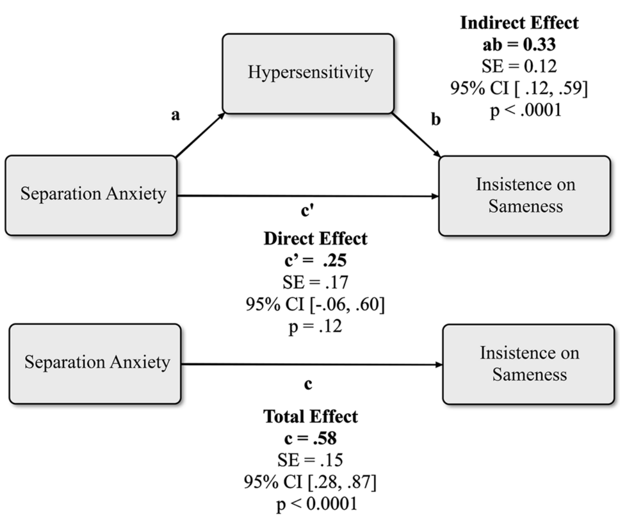


Fig. 3 Mediation model testing the effect of hypersensitivity on the relationship between separation anxiety and I/S (ASD group)

Overall, separation anxiety was related to I/S (c = 0.58, SE = 0.15, 95% CI [0.28, 0.87], p < 0.0001). The indirect pathway from separation anxiety to I/S through hypersensitivity was significant (ab = 0.33, SE = 0.12, 95% CI [0.12, 0.59], p < 0.0001), with hypersensitivity mediating 57% of the total effect. However, the direct path accounting for the contribution of separation anxiety to I/S, independent of hypersensitivity, was not significant (c' = 0.25, SE = 0.17, 95% CI [−0.06, 0.60], p = 0.12).

Two additional models were examined for the three variables, testing alternative mediators in the place of hypersensitivity. The first of these included separation anxiety as a mediator between sensory hypersensitivity and I/S, and was not significant (ab = 0.04, SE = 0.03, 95% CI [−0.01, 0.11], p = 0.12). The second included I/S as a mediator between sensory hypersensitivity and separation anxiety, and was also not significant (ab = 0.08, SE = 0.05, 95% [−0.03, 0.16], p = 0.11). These additionally tested models provide evidence for the specific directionality of the relationships described in our original model (Fig. 3).

Discussion

The goal of this study was to determine the impact of the relationships between hypersensitivity, anxiety subtypes (specific phobia, separation anxiety, and social anxiety) and I/S for groups of autistic and typically developing children. Our findings demonstrate that, in autism, specific phobia and separation anxiety are related to I/S because of their co-occurrence with hypersensitivity. Put differently, hypersensitivity in autism strongly mediates the relationships between (a) specific phobia and I/S, and (b) separation anxiety and I/S. Importantly, we observed this relationship in autistic, but not TD children. No evidence was found linking hypersensitivity and social anxiety in either group, although social anxiety was correlated with levels of I/S in autism.

Our mediation results provide novel support for directional relationships between anxiety, hypersensitivity, and I/S in autism. These findings may explain previous discrepancies in studies administering measures of total anxiety (Lidstone et al. 2014; Wigham et al. 2015). Total anxiety may provide a general understanding of the relationship between anxiety and RRBs, however, our data clearly suggest that different subscales of anxiety have distinct relationships with I/S and hypersensitivity. Both specific phobia and separation anxiety exhibited a significant relationship with I/S, which was mediated by hypersensitivity. Social anxiety, on the other hand, showed no relationship with hypersensitivity. This highlights the importance of separating measures of total anxiety into specific components.

The mediating role of hypersensitivity on specific phobia and separation anxiety in the ASD population has, to our knowledge, not been previously reported. In non-ASD adult samples, I/S has been related to panic disorder (Boswell et al. 2013) as well as separation anxiety (Boelen et al. 2014). However, hypersensitivity was not explored in either of these investigations. In regards to specific phobia, we postulate that this relationship could be due to repeated exposure to everyday sensory experiences that do not induce to anxiety in non-ASD children. Such experiences may elicit a physiological stress response in a population experiencing hypersensitivity, with I/S as a resultant behavioral coping mechanism. For example, an autistic

child's experience of a shopping mall with loud music, bright displays, and intense smells from the food court or beauty shops, may result in a "sensory overload" which overwhelms the child's ability to cope. Thus, the relationship between how "anxious" the child is in a given environment and their behavioral symptoms (i.e. I/S) is greatly impacted by their sensory experience of the event. Future work is needed to explore this and other possibilities. The relationship between separation anxiety, hypersensitivity, and I/S has been less studied. Although there is a dearth of research on the topic, our predictions were based on one study showing a significant correlation between separation anxiety and I/S (Boelen et al. 2014). These results provide ample reason to explore these relationships in future work.

New findings suggest that intolerance of uncertainty (a construct highly related to I/S) and associated anxiety may be adaptively addressed by helping parents to reduce avoidance strategies (Hodgson et al. 2016; Rodgers); likewise, the mediating effect of hypersensitivity observed in these data suggest that sensory sensitivities may also provide an appropriate target for remediation. From a clinical standpoint, there are two distinct approaches that could be useful in targeting sensory sensitivities as related to anxiety. These approaches depend upon whether these reported sensory sensitivities are *sensitivities* per se (i.e. lower sensory thresholds resulting in increased detection and saliency of a given sensory input) or if they are increased sensory *reactivity* (i.e. an atypically heightened behavioral response to a similar sensory perception). As it is a parent report, the Sensory Profile is unable to distinguish between the two. In the case of increased sensory *sensitivity*, it would make sense to try to lessen the intensity of the stimuli itself, which could be done by making accommodations to the environment (such as tinted glasses, special glasses for fluorescent lighting, or headphones/earplugs to reduce noise, etc.). Alternatively, in the case of increased sensory *reactivity*, an alternate approach might be to reduce the anxious physiological response to adverse stimuli through graduated exposure.

The absence of a mediational relationship between social anxiety and hypersensitivity in ASD (thus obviating any three-way relationship such as a mediation) was unexpected. With that said, it has been suggested that the prevalence of social anxiety in autism may in fact be almost two times lower than previously reported (Magiati et al. 2016; van Steensel et al. 2011), and these current data reflect that possibility. Although our initial finding that social anxiety and I/S were related for children with ASD was, in fact, contrary to two previous studies which reported a non-significant relationship between the variables (Magiati et al. 2016; Rodgers et al. 2012), others have suggested that the variable levels of anxiety reported in ASD may reflect a difference in how it presents in ASD relative to TD, as opposed to a difference in prevalence (Kerns and Kendall 2012). As there is a growing body of evidence linking social anxiety symptoms in autistic adolescents with increased levels of autism-related behavioral issues, this relationship is of clinical importance and should be explored further (Ambler et al. 2015; Pugliese et al. 2013; White et al. 2012).

Future Research

We examined a group of verbal autistic children with, on average, normal to high IQ scores. However, a small group of children in the sample presented with intellectual disability (ID; IQ < 70). While our N within this lower IQ range was too small to draw conclusions, in future studies, the mediation effect should be examined separately for groups of autistic children with and without ID, as it currently remains unclear whether hypersensitivity occurs more frequently and intensely for children without ID (Glod et al. 2015). Additionally, a number of researchers have reported that hypersensitivity peaks for autistic children between the ages of 6–9 years (Ben-Sasson et al. 2009; Green et al. 2012). We did not observe a significant correlation between hypersensitivity and age in autistic children. However, while our age range of 7–17 provided the range needed for this age analysis, our sample size at each age limited our ability to draw conclusions regarding specified age ranges. Conversely, we did find evidence of a positive correlation between symptom severity and hypersensitivity in our autistic group. This suggests that the intensity of hypersensitivity impairment may increase in tandem with overall autistic symptomatology—which has been shown to remain generally stable over time (Louwerse et al. 2015). Our study also relied exclusively on parent reports of child anxiety symptoms; although parent and child reports have previously shown good consistency, more recent recommendations indicate that a multi-informant approach incorporating child and teacher reports may further improve understanding (Weiss 2014). Furthermore, the scales used here were adapted to explicitly measure our constructs of interest in an ASD population, and an independent validation of these newly configured measures is needed before drawing stronger conclusions.

Finally, in terms of the TD group itself, significant relationships were not observed between anxiety, hypersensitivity, and I/S, making a mediating effect statistically impossible. With that said, while the TD group did not exhibit significant correlations between variables, the direction of these non-significant relationships were quite similar to the ASD group in most cases. TD data from each of these three variables were truncated relative to their TD peers, as would be expected, which may have contributed to these non-significant results. Indeed, many

measures find that ASD symptomatology shows similar, yet non-clinical, patterns in TD samples (Maisel et al. 2016; Mayer 2017) and sensory sensitivity has also been shown to be a contributing factor to the development and maintenance of affective disorders (Ahadi and Basharpoor 2010; Goldsmith et al. 2006; Neal et al. 2002). As such, these non-significant findings should not be interpreted as strongly claiming that no relationship exists in TD groups, and these relationships warrant further study.

Conclusion

Here, we have highlighted the critical importance of studying sensory hypersensitivity in autism in a manner that accurately captures the complexities of the multi-faceted disorder. Furthermore, our examination of effects in comparison to a group of typically developing children indicates that the mediational effect of hypersensitivity on anxiety subtypes and I/S is seen only in autistic children. Future treatment planning may be improved by addressing symptoms related to social anxiety with a different approach from those related to specific phobia or separation anxiety. If hypersensitivity is in fact at the root of these relationships, existing anxiety-focused behavioral interventions may lead to improved outcomes by including remediation and coping skills specific to reducing sensory sensitivity.

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